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LETTER TO THE EDITOR

Painful intraosseous leiomyoma of the distal femur

Dear Editor:

Intraosseous leiomyoma is a rare tumor usually arising in the axial skeleton. Involvement of the peripheral skeleton is even rarer with only five cases reported in the literature. Herein, we report a case of a 57-year-old male with intraosseous leiomyoma presenting as a painful swelling mass in the left distal femur. Initial computed tomography (CT) of the left knee revealed a 6.5-cm osteolytic lesion with mild cortical irregularity. Magnetic resonance imaging (MRI) showed a soft tissue tumor mass in the left femoral condyle. This unusual tumor location and size made differential diagnosis and surgical decision-making challenging. Herein, we discuss the clinicopathological features of this case and review the literature on primary intraosseous leiomyoma.

A 57-year-old man visited our hospital complaining of left knee pain that had lasted for several years. Physical examination showed painful swelling over the left femoral region but no limitation in range of motion. He had no fever and normal laboratory findings. Plain radiograph (Fig. 1A) and CT (Fig. 1B) revealed an osteolytic lesion (6.5 cm) with mild cortical irregularity. MRI showed a soft tissue tumor in the left medial femoral condyle and supracondylar region, manifesting iso-signal intensity to muscle on T1 weighting images, and iso to high-signal intensity on T2 weighting images with fat saturation. On analysis of imaging, the tumor could be a giant cell tumor, bone metastasis, or other rare entity.

Biopsy before the surgery or frozen section during surgery may be helpful for surgical decision-making. Analysis of the intra-operative frozen section suggested leiomyoma. Based on this diagnosis and considering the stability of the knee joint, *en bloc* resection and arthroplasty with prosthesis was performed. The specimen showed a well-demarcated gray white tumor, 7.2 × 5.5 × 4.5 cm, involving the epiphysis and adjacent metaphysis of the distal femur (Fig. 1C). No obvious destruction of the overlying periosteum and adjacent soft tissue could be identified macroscopically. Extensive sampling by taking blocks per centimeter was performed.

Histologically, the tumor was composed of moderately cellular spindle cells arranged in orderly intersecting fascicles, with abundant eosinophilic cytoplasm and elongated blunt ended "cigar-shaped" nuclei (Fig. 1D). The cellular atypia was inconspicuous. No mitosis or necrosis was found. Masson's Trichrome stain showed tumor cells with muscle differentiation. Immunostaining showed a diffuse and strong positive for smooth muscle actin (SMA) (Fig. 1D) and a focal positive for desmin. The absence of cellular atypia, mitosis, and necrosis in a smooth muscle tumor of bone was consistent with an intraosseous leiomyoma. Two years after the surgery, the patient showed neither loosening nor evidence of tumor recurrence in a plain radiograph.

Leiomyoma is a benign smooth muscle tumor found most commonly in the uterus, gastrointestinal tract, and skin. Due to the paucity of the smooth muscle in the bone, intraosseous leiomyoma is very rare with fewer than 30 reported cases. It is thought to originate from the smooth muscular tissue existing in the blood vessels of the bone. Involvement of the peripheral skeleton is even rarer with only five reported cases, originating in the ulnar, tibia, fibula, and femoral neck [1–5]. Our case was found in the distal femur, a previously unreported location for this entity.

Intraosseous leiomyoma involving the peripheral skeleton has been found in adults with an age range of 31–57 years and with a slight female predominance (Table 1). The reported tumors have ranged from 8 mm to 7 cm. Surgical methods vary depending on tumor size and location. Tumors arising from the peripheral skeleton usually present as painful lesions. On the contrary, most intraosseous leiomyomata arising from the skull bone were painless swellings on initial presentation.

Leiomyosarcoma of the bone is more common than intraosseous leiomyoma. Differentiating between the two is important and challenging. The histological criteria have not been well established due to the rarity of the tumor. We based our diagnosis on the absence of mitosis(cutoff point 4/50 high-power fields), cellular atypia, and necrosis, the criteria used in distinguishing between soft tissue leiomyoma and leiomyosarcoma. Further research and more clinical experience are needed to confirm whether these criteria can be used to distinguish between the two entities.

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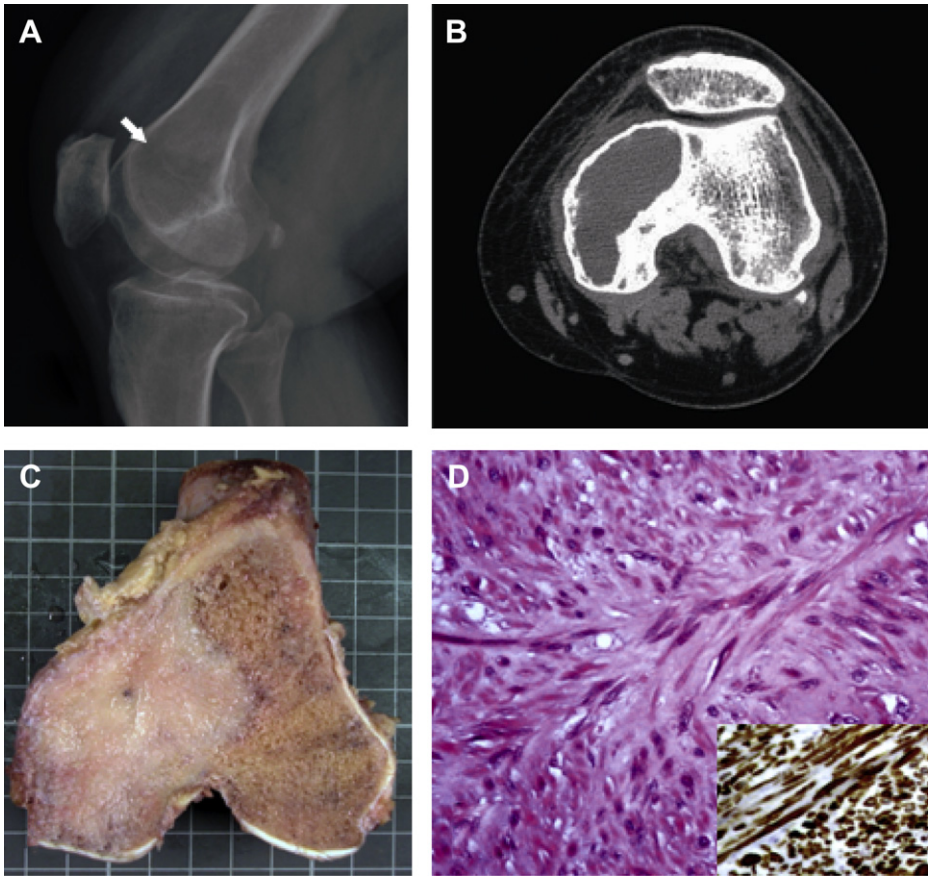

Figure 1. (A) Plain radiograph revealed an osteolytic (radiolucent) lesion (arrow) of the left knee. (B) Computed tomography (CT) showed a tumor with mild cortical irregularity. (C) Grossly, there was a well-demarcated gray white tumor, 7 cm in greatest diameter, involving the distal femur. (D) Histologically, the tumor was composed of bland spindle cells arranged in orderly intersecting fascicles, with abundant eosinophilic cytoplasm and elongated blunt ended nuclei (hematoxylin and eosin; original magnification, 400×). Tumor cells were diffuse positive for SMA immunostain (lower right panel; original magnification, 400×).

Table 1 Intraosseous leiomyoma involving the peripheral skeleton.

No.	Location	Age (y)/sex	Size	Clinical	Treatment
1	Fibular	42/Female	2 cm	N/A	Curettage
2	Ulnar	31/Female	8 mm	Painful	*En bloc* resection
3	Tibia[a]	37/Female	2 cm	Painful	Surgical excision
4	Tibia	37/Male	2 cm	Painful	*En bloc* resection
5	Femur neck	54/Female	N/A	Painful	Surgical excision
6	Distal femur	57/Male	7 cm	Painful	*En bloc* resection

N/A = not available.
[a] Leiomyoma of the periosteum of the tibia.

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